

Hepatic hydatid cyst rupture into biliary tree presented as anaphylaxis and obstructive jaundice

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Introduction:

Hydatid disease is a parasitic infection by a tapeworm of the genus *Echinococcus* it is a zoonotic infection. Echinococcosis is unusual in northern Europe, U.S.A. and the endemic areas are the middle east, as well as other part of the world, including India, Africa, South America, Turkey, Australia, New-Zeland and southern Europe 1-3.

The incidence of cystic echinococcosis is 1-220, cases per 100,000 population, while that of alveolar echinococcosis is 0.03-1.2 cases per 100,000, it is much more rare 4

The morbidity is due to free rupture or infection of the cyst or the dysfunction of affected organs such as biliary obstruction, liver cirrhosis, bronchial obstruction, and renal out-flow obstruction⁴, and increase Intracranial pressure due to mass or hydrocephalus.

The mortality is due to anaphylaxis, systemic complication of the cyst (sepsis, cirrhosis, respiratory failure) or due to operative complications.

The mortality rate is 50-60% in alveolar form of echinococcosis, and may reach up to 100% in untreated cases.

Sudden death has been reported in asymptomatic form of this disease. Infestation by hydatid disease in human most commonly occurs in the liver 55-70% followed by the lungs (18-35%). The two organs can

be infected simultaneously in about 5-13% of cases 5,6.

Even though hydatid disease can occur in any organ⁷, it is rare to be seen in organs like brain, heart, pericardium, spinal cord, and eye ball. Spontaneous rupture of the cyst into hollow viscera is an extreme complication with an estimated frequency of 0.5% leading to hydatidemia and hydatid-diarrhea 8,13.

Although hydatid cyst of the liver is frequently encountered, its rupture into biliary tract causing obstructive jaundice with anaphylaxis is an uncommon presentation.

The aim of reporting this case is that in endemic areas in patients with anaphylaxis and/or obstructive jaundice, hydatid disease should be suspected and considered in the differential diagnosis.

Case report:-

An 18 years old female, from Sana'a non-smoker, she is not chewing khat, illiterate, married for one year, but has no children,

She was presented with abdominal pain, vomiting, and dizziness, followed by unconsciousness, associated with swelling of eye-lids and lips, her past medical history is unremarkable, No history of drug intake, insect bite, No H/O Allergy, Eczema, Asthma or Rhinitis.

Her family history is irrelevant. On examination: patient was unconscious with erythematous skin and edematous lips and eye lids and was in a state of shock. After resuscitation, she was admitted to the ward, and her systemic examination revealed huge-hepatomegaly.

Three days later the patient started to have wheals and deep jaundice.

Her investigations showed Hb 13gm WBCs 16.4 x1000/mm³, 4% eosinophilia plat. 195,000 ESR 22 Other investigations were all normal. In the 4th day her liver function tests showed ALT 80, AST 72, ALK .Ph. 524, total bilirubin 1260 μ mol/L. prothrompin time was 19 sec.

Her abdominal ultrasonography is shown in (Fig.1) and C.T. scanning (Fig.2) a big cyst in the right lobe of the liver

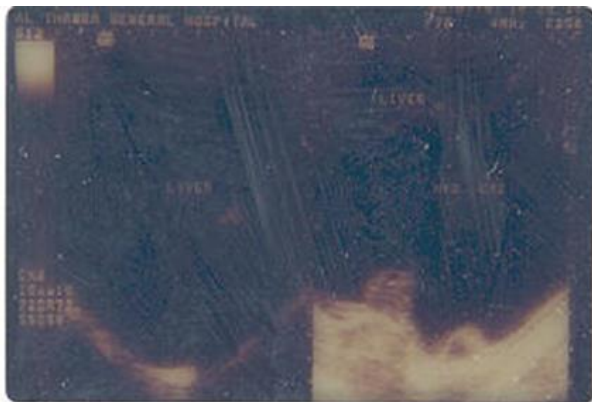
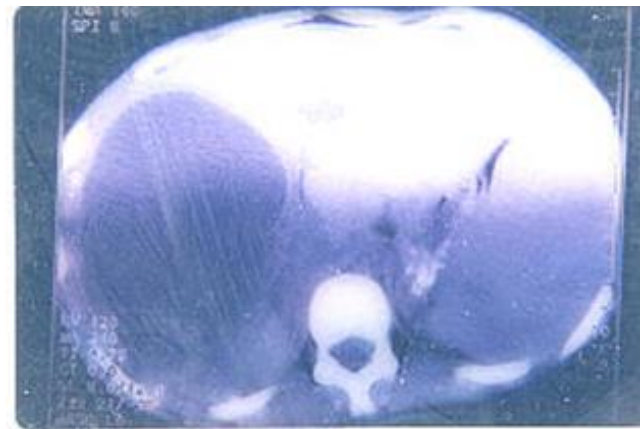
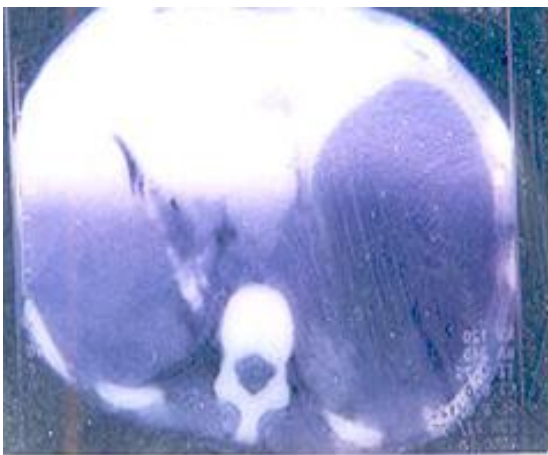


Fig. (1) Ultrasound of the abdomen showing a big cyst in the right lobe of the liver



Discussion:

Yemen is one of the areas where hydatid disease is not rare in the population and can affect any organ in the body but the liver and the lungs are the most frequently affected organs. Hepatic hydatid disease is the commonest form of echinococcosis. The right lobe of the liver is affected in 80% of the cases and the left lobe in 20%. Generally complications of hepatic hydatid are uncommon but when they occur, the commonest is rupture into the biliary tree. It occurs into the right duct in 55-60% of cases, into the left duct in 25-30%, and rarely into the confluence of gall bladder [14,15]. Intrabiliary rupture can lead to obstructive, septic, or allergic manifestations. Patients commonly present with right abdominal pain (82%), obstructive jaundice (57-100%), fever (70-90%), acute cholangitis (20-30%), abdominal lump (22-39%), and rarely with

acute pancreatitis, liver abscess or septicemia,, or it may be asymptomatic (5-6%).^{16,17}

Rupture of hydatid cysts have been classified into three types by Lewall and McCorkell¹⁸. Contained, communicating and direct. Communicating rupture can be simple communication between small biliary radicles and the cyst, or frank rupture into the biliary tree (as in this case). An accurate pre-operative diagnosis helps in planning the line of treatment and also helps during surgery. Ultrasound and CT are routinely used to confirm the diagnosis, but rupture of the cyst and evacuation of its contents may change the radiological appearance necessitating more sophisticated investigations like MRI, MRCP, ERCP and hepatobiliary scintigraphy¹⁹.

The major complications of hepatic hydatid disease is death due to anaphylaxis, infection with septic syndrome, cystobiliary fistula and obstructive jaundice.

The clinical presentation of hydatid disease depends on size, site of the cysts and accessibility of the organ involved for clinical examination.

In our community accidental discovery of the disease is frequently observed during investigations of patients for unrelated disorders. Eosinophilia is expected to be found in 25% of hydatid disease but it is not seen in this case. Preoperative diagnosis of hydatid can be made by ultrasonography and confirmed by C.T. scanning.

Both are the most helpful ways in our hospital. Other techniques are helpful in other sites like MRI in brain and echocardiography in cardiac hydatids^{9,10,20}.

Serological tests which are used for diagnosis are varied and have been evaluated by various authors with variable results. They are being carried out for diagnosis, screening and post operative follow up for recurrence.

The indirect hemagglutination test and the enzyme-linked immunosorbent assay (ELISA) have a

sensitivity of 80% overall (90% in hepatic and 40% in pulmonary) and are the initial screening of choice.

Immunodiffusion and immunoelectrophoresis demonstrate Antibody to Antigens and used for confirmation²¹.

The treatment of hydatid cyst is principally surgical, however pre-and postoperative one month course of Albendazole with or without two weeks of Praziquantel should be considered in order to sterilize the cyst, decrease the chances of anaphylaxis, decrease the tension in the cyst wall so reducing the spillage during surgery, and to reduce the risk of recurrence rate postoperatively^{1,7,21}.

The dose of Albendazole is 10-15 mg/kg daily and that Praziquantel is 40mg/kg per week^{1,4,7,21}.

Intraoperatively the use of hypertonic saline or 0.5% silver nitrate, before opening the cavities, tends to kill the daughter cysts therefore prevent further spread or anaphylactic reaction²¹. Other scolicidal agents include formaline, hydrogen peroxide, absolute alcohol, and chlorhexidine.

The direct mortality due to hydatid cyst is very low ranging from 0.29-0.6%, while the indirect operative mortality is 0.5-4%.⁴

In conclusion: hydatid cyst has been reported in any part of the body, no site is exempted, and it should be thought of, in an endemic area in any cystic lesions or unexplained anaphylaxis. The latter complication could be seen even in normal looking unruptured cyst, due to microscopic rupture.

The elective surgical technique should precede and be followed by medical therapy.

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